to cause a predisposition to fatty infiltration of the liver. It is to the acute infections that the change in this series of cases must be ascribed.

Cirrhosis. One liver was the seat of hypertrophic cirrhosis. It was the case of a female infant, three months old, with a typical syphilitic eruption, fissured lips, and snuffles. She was fairly well nourished, and entered the hospital for vomiting and diarrhœa. The liver extended 3 cm. below the free border of the ribs, and the spleen was also enlarged. There was no icterus at any time. At the autopsy the liver weighed 205 grammes, was decidedly enlarged, and cut with difficulty, owing to the presence of an increased amount of connective tissue. Microscopically there was a decided increase in the interlobular connective tissue and less of the intralobular. This connective tissue was partly of the cellular, and partly of the older, fibrous variety. The liver cells were granular, and, in places, the seat of pressure atrophy. The gall-ducts were normal in structure and not increased in number. The walls of the small bloodvessels were thickened, the adventitia and the intima being involved. The spleen weighed 150 grammes; it was firm, and the capsule showed chronic perisplenitis. Microscopically hyperplasia was marked.

The only other case of cirrhosis encountered in this series of autopsies was one due to obstruction of the bile-ducts in an infant dying at the age of three months. It is reported elsewhere.[1]

No case of waxy degeneration occurred in these young children.

THE SACROCOCCYGEAL DIMPLES, SINUSES, AND CYSTS.

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The purpose of this paper is to present a preliminary consideration of the congenital dimples, sinuses, and dermoid cysts found in the median line of the back over the coccyx and lower part of the sacrum, together with a more or less complete review of the literature of the subject to the year 1901.

The presence of these congenital defects occurring in a far larger number of cases than is ordinarily believed has received as yet comparatively little attention from clinicians, having been remarked only as a rule when some surgical complication, to which the deeper of these structures (the sinuses and cysts) are especially liable, has made a careful examination necessary.

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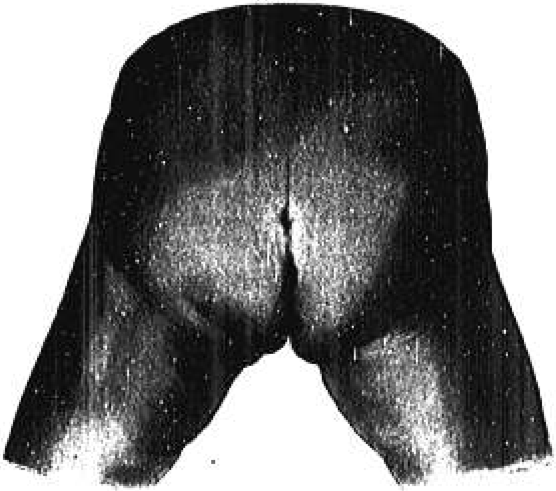
[1] Archives of Pediatrics, March, 1902.

The French authors were the first to regard the differences in development of the sacral region of the trunk, and, therefore, to note those longitudinal furrows or rounded depressions of greater or less depth, found in a large proportion of babies and children and smaller proportion of adults.

A simple examination of adults or children, but especially of babies, by the mere separation of the buttocks, will often show over the coccyx above the anus, some form or degree of dimpling or indentation of the skin, either the longitudinal furrowing or rounded depression, with less frequently a sharply marked deep pit indicating the mouth of a fistula or sinus. (See Figs. 1, 2, and 3.) Nor is it very uncommon to find several members of one family showing the same condition. Such cases have been reported by Madelung and Dunlop.[1]

FIG. 1.



Representing the most common appearance, the shallow dimple. Drawn from ten-days-old baby. Corresponds to A, Fig. 4.

The integument within and immediately about these little defects usually shows slightly more gloss, is pinker than that over the rest of

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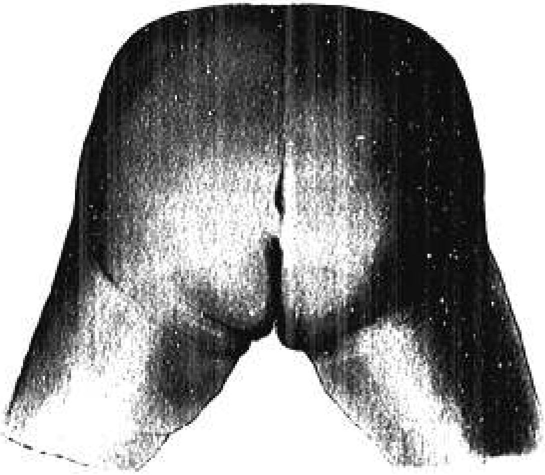
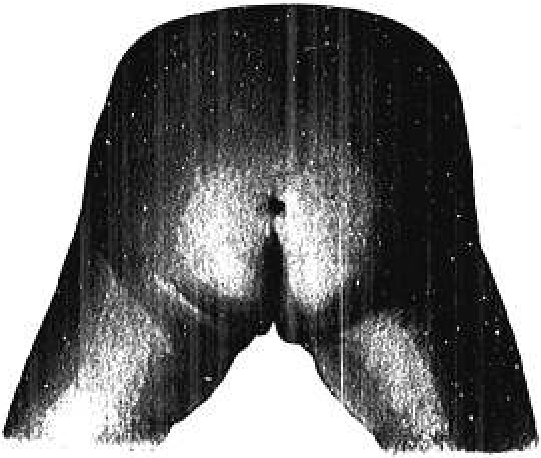
[1] Lancet, London, 1882.

FIG. 2.



Representing the longitudinal furrowing. Drawn from baby about one week old.

FIG. 3.

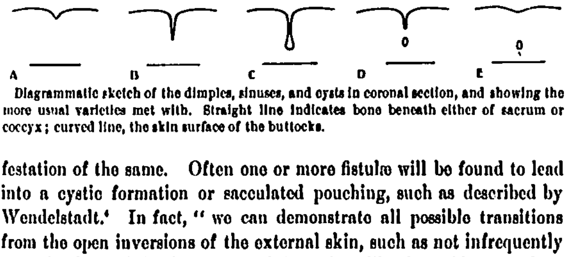


Representing the external appearance of the less frequently found sinus. This may be only a centimetre deep or several centimetres, and may or may not be sacculated at its end. Corresponds to the external appearance of B, C, or D, Fig. 4.

the part, and is moister. From the deeper ones, indeed, clothing in contact with the skin may be decidedly dampened, as instanced in some of the cases of J. M. Warren[1] and others. Pulling upon the skin about these depressions or putting it upon the stretch with the finger demonstrates more than the usual resistance to its gliding on the deeper structures, and at the same time causes an exaggeration of the dimpling, showing an adherence or tacking down to the less movable parts beneath. Hodges,[2] among others, has written of this. It can often be shown where there is but the merest dimpling present, one so slight as to escape ordinary observation. It is to be regarded as quite characteristic of these depressions.

In size and depth they may range from the faintest indentation through sharper and deeper pit-like formations to sinuses of many centimetres depth, possessing the finest orifice or one so large as to be mistaken for the anus itself, as happened in a case reported by Lannelongue.[3] Or we may find over this sacrococcygeal area a completely sequestrated pouch, an epidermoid cyst, with or without outward manifestation of the same. Often one or more fistulæ will be found to lead into a cystic formation or sacculated pouching, such as described by Wendelstadt.[4] In fact, "we can demonstrate all possible transitions from the open inversions of the external skin, such as not infrequently occur in the neck in the form of cleft or sinus-like dermoids, to such as have only a very narrow communication with the skin, and finally such as are wholly cut off, as was first demonstrated by Heschl."[5] (See Fig. 4.)

FIG. 4.



Diagrammatic sketch of the dimples, sinuses, and cysts in coronal section, and showing the more usual varieties met with. Straight line indicates bone beneath either of sacrum or coccyx; curved line, the skin surface of the buttocks.

The occurrence of these congenital defects observed in a study of a great number of subjects shows the largest proportion to exist in young babies, and the least in adults. Lannelongue gives 20 to 25 per cent. of all young children. Despres,[6] 30 to 33 per cent. at birth. Heurtaux,[7] 4 to 5 per cent. of adults on a basis of 960 cases examined.

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[1] THE AMERICAN JOURNAL OF THE MEDICAL SCIENCES, 1854; Philadelphia Medical Times 1873; Boston Medical and Surgical Journal, 1877, and "Surgical Observations."

[2] Boston Medical and Surgical Journal, 1882.

[3] Bull. Méd., Paris, 1889; Bull. Mém. Soc. de Chir., 1882-1886.

[4] Dissertation, Bonn, 1865.

[5] Prager Vierteljahrsschr., 1860.

[6] Bull. de la Soc. Anat., 1874; Bull. et Mém. Soc. de la Chir., 1889, vol. iv.

[7] Bull. et Mém. Soc. de Chir., 1882.

Lawson Tait,[1] 23 per cent. of women examined at the Birmingham Hospital for Women, which would certainly seem a high percentage for adults. Peyramaure-Duverdier,[2] about 3 per cent. of persons examined. Kuhn and Wendelstadt give a somewhat higher percentage. In a series of 300 consecutive births observed in this city, a well-marked dimple or sinus occurred in 100 out of the 300 babies. The series was an absolutely consecutive one, and therefore gives very accurately the true proportion of cases presenting these anomalies.

As regards their situation, it may be noticed anywhere from the anus to well up on the sacrum and always exactly in the middle line. Usually they are situated over the lower part of the coccyx, rarely closer than 1 centimetre to the anus. Mallory[3] gives the situation in order of frequency as:

1. Near tip of coccyx.
2. Sacrococcygeal region.
3. Over lower part of sacrum.
4. Between coccyx and anus.

Nicaise[4] says they occur in three principal places, dorsum of sacrum, sacrococcygeal junction, and tip of coccyx. Lannelongue found in 95 observations that 29 were situated over the sacrum, 38 at the junction of the sacrum and coccyx, and 28 at the tip of the coccyx. Despres believes that they are commoner between the coccyx and the anus, while Lawson Tait believes he has seen the greater number at the upper end of the fold between the nates over the sacrum.

In the series here reported the average distance was 1.66 centimetres above the anus, agreeing more closely with the statistics of Mallory. The nearest to the anus was 1 centimetre, the most distant 3¼ centimetres.

There is more often but one dimpling or sinus noted, but occasionally two, more rarely three or more. Lannelongue reports those with three openings. Lamadrid mentions one with three openings, while Heurtaux reports one with three and another case with four congenital openings of sinuses in the middle line of the back. One depression was found ninety-two times, two depressions eight times. Those with three or four were not noted in the above series.

The "depression" of the French, or dimple, or fovea, is by far the most frequent and simplest of the three abnormal formations, *i. e.*, foveæ coccygeæ, fistulæ coccygeæ, and cysticæ coccygeæ.[5]

The sinus is much less frequent. In the 100 cases referred to the foveæ occurred eighty-nine times, while the fistulæ occurred but eleven times. Of the relative proportion of the cyst occurrence we can as yet say nothing for lack of sufficient data and histological examination.

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[1] Publication, Dublin.

[2] Thèse de Paris, 1882.

[3] THE AMERICAN JOURNAL OF THE MEDICAL SCIENCES, 1892.

[4] Tribune Médicale, 1893.

[5] Wette's Classification, Langenbeck's Archiv, 47.

By the older writers, before the days of microscopical examination and before any serious attempt at explanation for their occurrence was made, but when the surgical complications of certain of these structures formed subject-matter for literature, they were usually described as "pilous cysts," or "pilonidal sinuses;" or they were styled "furuncles," "abscesses," or "phlegmons," according to the nature of the pathological process occurring and in which they were seen.

The study of these foveæ, fistulæ, and cysticæ is of comparatively recent date, beginning about the year 1868. Courand,[1] reviewing the literature of the subject to the year 1883, declares that he found but fourteen reported cases.

The tumors of this region, on the other hand, were studied many years previously. Meckel and Himly, in 1818, wrote upon fœtal inclusions and parasitic monsters. Ammon, in 1842, Wernher, in 1843, and Velig, at Strassbourg, 1846, Lotzbeck, in 1858, etc., contributed to the literature. One of the first reported cases of sinuses was that of Anderson,[2] in 1847, under the heading, "Hair Extracted from an Ulcer."

Gross,[3] in 1882, speaks of a "congenital pilocystic tumor" sometimes met with upon the coccyx in the vicinity of the anus. "Its origin is apparently connected with a sebaceous follicle, which, during its development, intercepts a small tuft of hair very much as in certain sebaceous formations on the head and face." Vaughan,[4] in 1865, considers their origin involved in obscurity. He reports three cases of "pilous cysts." Lamadrid,[5] 1872, considers them due to imperfect coalescence of the lateral halves of the body. "It is loose hairs which work their way down these preformed sinuses, which cause the irritation and inflammation often observed." J. M. Warren, in his *Surgical Observations*, also considers their origin obscure. Hodges, in 1880, believed as did Lamadrid, that hairs from the surface were the primary source of trouble. Heurtaux, in 1882, believes that epithelial detritus and sebaceous accumulations take place in these congenital infundibula. "This retention can become the cause in certain cases of suppurative inflammation of the skin which forms the bottom of the sinus; the skin becoming ulcerated and perforated, pus gradually infiltrates the subcutaneous tissue and gives rise to the fistulous tract."

What bothered the older observers was the presence of the coil of hair or accumulation which they usually found at the bottom of the sinuses and sacculated dilatations in their suppurative cases. As the epithelial, sebaceous, and hair-bearing lining was destroyed by the inflammation and often impossible of identification, they had to imagine the strands of hair working down from outside, quite in such a way "as hair-balls are formed in the stomachs of cattle," one writer states. Hodges gives three conditions as essential, "a congenital dimple or sinus, pilous development, and lack of cleanliness."

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[1] Thèse de Paris, 1883.

[2] Boston Medical and Surgical Journal, 1847.

[3] System of Surgery, vol. ii.

[4] St. Louis Medical and Surgical Journal, 1865.

[5] Philadelphia Medical Times, 1873.

If no hair was found it was the sebaceous and epithelial accumulation, with little or no means of exit, that caused the trouble. Walker,[1] in 1875, reported a case of "Monstrosity by Inclusion." A sinus over the coccyx was laid open and a coil of light hair found therein. Fere,[2] in 1882, thinks the dimples and sinuses due to incomplete closure of the "posterior umbilicus," *i. e.*, the lower end of the medullary tube. Terrillon,[3] in 1882, considers them due to a special form of spina bifida involving the skin only. Lawson Tait, suggested the "coccygeal dimple" as the "cicatrix of the spina bifida by which the human tail has been lost." Lannelongue, in 1882, believes that "after the medullary canal is formed, the mesoblast passes back between the vertebral column and the external epidermis, except in the region of the sacrum, where little of this tissue is interposed, so that this region is reduced to epidermis and bone. Consequently the superficial layer, the epiblast, joined at a later period to the mesoblast, preserves closer relations with the bone, and at one or more points the skin may be bound down to the bone, and later, when the subcutaneous tissue is developed around these places, a depression will be formed. If deep and narrow enough, the orifice may close up and a dermoid cyst will be the consequence." Wendelstadt, in 1885, expresses the same idea and opinion.

As these congenital dimples and sinuses were not confined in their occurrence to the sacrococcygeal region, but were not infrequently observed in the same localities as the dermoid cysts of regions in which embryonal parts joined, such as the neck, face, roof of the mouth, back of the head, and along the spine, and even in conjunction with these cysts (see the cases of Guzman,[4] Sutton,[5] Stelwagon,[6] Barling,[7] Wagstaffe, etc.), the idea of imperfect closure of the lateral halves of the body or epiblastic involution was usually adduced to explain them. This was the generally accepted explanation up to the time of the article by Tourneux and Herrmann,[8] in 1887, upon the remains of the spinal cord or medullary vestiges in this region.

Tourneux and Herrmann, finding remnants of the spinal cord rudiment behind the coccyx in the fœtus, in a study of this region, believed them of sufficient importance to account for these structures. Borst[1] gives the salient features of the argument: "At the beginning of the third month the spinal cord reaches to the third last coccygeal vertebra and is continued to the lowest coccygeal vertebra as a bundle of irregularly arranged nerve fibres in which the cells are embedded in groups and rows apparently without distinct regularity, and there is no central lumen. This bundle of fibres is connected with the deep layers of the cutis at the lower end of the spinal column, and these structures form a swelling and a cavity, the latter partly lined with polyhedral epithelium and partly with a form more on the cylindrical order. Owing to the unequal growth of the outer coverings of the spinal column and of the spinal cord, the growth of the soft parts being slower than that of the spinal column, a loop-like formation of this terminal portion of the spinal cord gradually develops, which bends toward the dorsum of the fœtus (horseshoe form of the terminal end).

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[1] Virginia Medical Monthly, 1875.

[2] Bull. et Mém. Soc. de Chir., 1882.

[3] Bull. de la Soc. Anat., Paris, 1878-1882.

[4] Thèse de Paris, 1883.

[5] Journal of Anatomy and Physiology, London, 1886; Annals of Surgery, 1889; Lancet, London, 1889.

[6] Philadelphia Medical Times, 1883.

[7] Annals of Surgery, 1889.

[8] Journ. de Anat. et Phys., Paris, 1887.

"The concavity of this loop is directed backward and upward; the two ends of the loop terminate, the one anteriorly and deep seated (descending), and the other posteriorly and superficial (ascending), the latter lying immediately beneath the skin in the deep tissues behind the last coccygeal vertebra. At the end of the third month the conus of the spinal cord is on the level of the junction of the third and fourth sacral vertebra, and below this there is only an epithelial tube surrounded by a thin layer of longitudinal nerve fibres, which structure broadens along the lower coccygeal vertebræ (the descending end) and terminating below in the ascending end as mentioned above; the filum terminale developing out of the upper sacral portion of this cord, while the lower gradually disappears. So the so-called 'ascending' of the spinal cord is not only due to an unequal growth of the spinal cord and spinal column, but also to the fact that a portion of the former undergoes retrograde changes and disappears.

"The deeper end of the before-mentioned loop, as intimated, undergoes retrograde changes by atrophy during the fourth month. But the upper end of the mentioned loop, which at this time lies over the second last coccygeal vertebra under the skin, continues to grow up to the fifth month, then reaching its maximum development (⅜ millimetre along the fourth and fifth coccygeal vertebræ); in the formation of strands and heaps of polyhedral cells with irregular cavities lined with cylindrical, cuboid, or polyhedral epithelium, the whole extending from below anteriorly to above posteriorly, and connected with the tip of the coccyx by means of the so-called caudal ligament; this ligament accompanies the entire medullary remnants throughout their whole extent.

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[1] Centralblatt. f. Allgem. Path. u. Path. Anat., 1898.

"These mentioned medullary remnants also begin to atrophy after the fifth month; but traces of the same, it is said, can be found in the fully developed newborn.

"At the middle of the sixth month only two or three of the mentioned heaps of cells remain, each possessing a cavity, and in the wall of the latter small, round cavities are found between the cylindrical or flat cells.

"Of the cells lining the central cavities of these structures (Perman[1]) some become flat, resembling squamous epithelium (as that of the superficial skin), which might indicate a return to the former ectodermal type; others become elongated and resemble the cylindrical cell of the ependyma.

"The cells *forming* the mass of the medullary remnant retain their spherical or polyhedral shape. Ciliated epithelium has not been found in the medullary remnants." These researches introduced a new element in the discussion, and the persistence of these structures was considered by many as productive of sinus and cyst formation in this region.

Sutton, in 1889, considers it more satisfactory to regard these sinuses and cysts as "slight defects in the coalescence of the superficial portions of the medullary folds in the sacrococcygeal region."

Masse,[2] in 1889, agrees substantially with Lannelongue. Mallory, in 1892, working independently of Tourneux and Herrmann, after an examination of the bodies of six fœtuses by section through the sacrum and adjacent tissues, confirms their results. He attributes everything in the way of dermoid structure found over the coccyx and sacrum to these medullary remnants, neural vestiges, or "vestiges coccygiennes" of Tourneux and Herrmann. "These cases show," he says, "that in fœtuses of three to six months there is frequently present over the coccyx a canal lined with epithelium, in some cases connected with the skin, in others not; in some, situated near the skin; in others, near the coccyx. The question naturally arises as to their origin. They may be due either to an extension inward of the epidermis or to the remains of some canal. If due to an extension, or as Lannelongue assumes, to the skin being bound down to the coccyx, why do they not contain the glands and hair follicles with which the epidermis in that region is studded. As regards the extension inward of the skin, why should it occur here so often and nowhere else? It seems much more likely that they are due to incomplete obliteration of a former canal, and extending, as they all do, upward and posteriorly to the coccyx, the medullary canal seems to be the most likely origin.

"The branchial clefts are closed by the eighth week. As before stated, the medullary canal has been open as late as the ninth week, consequently the obliteration of the clefts in the one case, and of the medullary canal in the other, must take place at about the same period of intra-uterine life, with this difference, that the growth is more rapid and perfect in the upper part of the body, and hence more favorable to the closure of the clefts. If, notwithstanding this, sinuses and cysts occur in the neck and about the ears, there is at least an equal chance that they may occur at the lower end of the medullary canal.

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[1] Langenbeck's Archiv, 1895.

[2] Bull. et Mém. Soc. de Chir., 1889; Bull. gén. de Thérap., 1885.

"It would seem, from a study of the sections from these fœtuses, that obliteration of the medullary canal takes place at first and most completely at the lower end of the sacrum, and extends from this point in both directions. As is well known, the spinal cord at first extends the whole length of the vertebral canal; but, as the latter grows the more rapidly in length, the cord rises and the filum terminale is stretched, thus favoring obliteration of the medullary canal at the lower part.

"The obliteration of the medullary canal between the end of the vertebral canal and the skin apparently frequently takes place in an irregular manner; but, for that matter, the medullary canal in the spinal cord shows frequent irregularities, sometimes existing as a distinct canal, sometimes double, as in Fœtus VII., and often showing in sections only as a very irregular clump of cells. Undoubtedly, the majority of these remnants of the medullary canal become obliterated; only the larger, especially those in which glands and hairs are present, persisting as the depressions, sinuses, and cysts of extra-uterine life; and in all probability it is only the congenital sinuses and cysts which give rise to the suppurating sinuses."

Nicaise, in a clinical lecture reported in 1893, stated that his belief in the origin of these affairs agreed with that of Lannelongue and the majority of the French and German observers. This idea of epiblastic evolution, he says, was mentioned as far back as 1852 by Verneuil.

Ritschl,[1] in 1892, holds extreme views, believing that not only cysts situated upon the dorsum of the sacrum and coccyx, but cysts lined with stratified squamous epithelium, resembling that of the skin, and situated upon the ventral surface of the coccyx as well, are due to these same medullary remnants of Tourneux, Herrmann, and Mallory.

Aschoff,[2] in 1895, believes that the median agglutination may be the cause of sinuses and dermoid cysts of the dorsum of the sacrum and coccyx. He also says that the caudal extremity of the spinal cord lying on the dorsum curves upward and backward, forming a U-shaped loop, one end of which is connected with the skin. Gradually the loop portion which connects the ends disappears, and at the last the dorsal portion (les vestiges coccygiennes of Tourneux and Herrmann) does the same. The point at which connection with the skin exists, and which

[1] Beitrage zur klin. Chir., 1892.

[2] Cysten in Lubarsch's Ostertags Ergeb., 1895, vol. ii.

is connected with the coccyx by fibrous strands, is the fovea coccygea. This idea is again referred to by Stolper.[1] Borst, in 1898, is of the opinion that not only the depressions and sinuses, but the cyst formations and many of the more complex tumors of this region are due to these invaginations and agglutinations of the ectoderm. He believes with Lannelongue, Wendelstadt, Aschoff, and others, that the lower portion of the spinal column being the last portion of the posterior opening of the spinal canal to close, that, therefore, the "separation between the external skin and incurved corneal layer of the blastoderm is more difficult, and fibrous agglutinations with the skeleton are more apt to occur." That these fistulæ and depressions are not always connected with the spinal column by the fibrous bands mentioned by Fere and subsequently by Lannelongue (who denies this as an invariable rule), Wendelstadt, Molk,[2] Trendelenburg, and others can be explained upon the basis of their "subsequently becoming looser." He finds that these congenital depressions, sinuses, and cysts of the sacrococcygeal region "show the presence of the external skin in their walls." It is true, he says, as Wette[3] notes, that there are frequent deviations from the normal skin structure, such as thinning of the epithelial covering, absence of the development of papillæ, lack of true corium, poor development, or entire absence of hair, sebaceous and sweat glands; but this is no more than might be expected with such disturbances in development. Furthermore, due to complex conditions of development of this part of the body, these abnormal agglutinations can and often do occur, and "portions of the ectoderm become displaced beneath the surface, and there continue to develop."

Born, he says, having gone over all the work done in this line (Nagel, Mihalkovicz, Keibel, Reichel, Retterer, Tourneux, etc.), states that with the manifold and complicated processes of invagination and agglutination which occur here, epithelial germs can easily become separated and lead to the formation of cysts. Nor is it astonishing that cysts may occur in the perineal region as well, lined either with cylindrical epithelium or epidermis, as the ectodermal plate is able to produce epidermis as well as mucous membrane.

Stolper, in 1899, believes that Mallory and others "go far beyond the indications" in ascribing to the medullary vestiges the formation of these sacrococcygeal defects. "I am of the opinion that for the formation of fistulæ and true dermoid cysts we must also drop the frequently urged theory, or, at least, modify it very much. Particularly in these structures we miss the presence of nerve tissue as a principal constituent." The idea is a less radical one; it is not the medullary vestiges themselves, "but the peculiarities of growth associated with their development and atrophy are of interest in the development of fistulæ and cysts." It is the connective-tissue strands accompanying the ascending arm of the U-shaped tube, and which extend from the tip of the coccyx and later form the ligamentum caudale, which "are not without significance in the development of the mentioned depressions and furrows." "Even if the extra-vertebral nerve mass disappears after birth in the physiological way, the easily imagined bilateral continued growth of the accompanying connective-tissue strands may result in the formation of depressions and furrows, which, owing to comparatively early intra-uterine closure of overgrowing epidermis may result in canals. This becomes all the more probable when we consider that the medullary remnants have reached their maximum development at the beginning of the fifth month and begin to atrophy after the sixth month." He believes the epidermoid fistulæ and the sacral dermoid cysts to be a "continued development of the much more frequent and similarly explained foveæ coccygeæ." Both the fistulæ and the cysts are structures of true epidermis and with an epidermoid lining which is continuous with that of the external skin, and both are connected beneath with either the sacrum or the coccyx.

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[1] Deutsche Zeitschrift f. Chir., 1899.

[2] Thèse de Strasbourg, 1868.

[3] Arch. f. klin. Chir., 1891.

Wette's observations upon the often found relatively incomplete development of the skin lining these structures are also referred to. Owing, however, to the known irregularities in the formation of hair and the hair follicles and sebaceous and sweat glands in the different portions of the external skin, we should not be surprised to find similar irregularities in these complex malformations as seen elsewhere. Two cases are reported in which histological examination of the tissue removed was made. The first case, that of a man, aged thirty-one years, and suffering from an acute inflammation of one of these fistulous tracts (Clinic, Dr. Wagner, Konigschutte) was cured by radical excision of the whole tract. "It was found at several points that the wall of the fistula presented almost exactly the same histological structure as the external skin, except that the layer of epidermis was not so thick and had less of a corneal layer." There were no hair follicles found, but glandular structures very closely resembling sebaceous formations lay deeper in the skin than they are usually found. Disregarding the lesions of the acute inflammatory process and those due to previous curettings, "we had to deal with an open canal, open below, closed above, the wall of which was composed of epidermis throughout." The second case was that of an acute inflammatory process in a congenital sac showing similar conditions. Cure also resulted after radical extirpation. Microscopical examination of the sac here showed a mucous-membrane-like structure without hair follicles and with scant glandular structural development. The tract leading to the dilated portion was lined with granulation tissue, and no direct transition from the skin to the wall of the fistula could be made out, "but the undisturbed portion showed epidermoid lining."

This last case illustrates the almost completely sequestrated pouch, communicating with the exterior of the body by a narrow fistulous tract only. Some of these invaginations are at the start of considerable size, while others dilate from the closure of their fistulous outlets, the resulting accumulation often augmented by inflammatory products forming a tumor. This class must be grouped with the dermoid cysts.

We have, then, in this sacrococcygeal region three abnormal formations—depressions, sinuses, and cysts—usually communicating with the external surface of the body, sometimes not, and which are lined with a tissue identical with that covering the external surface, namely, skin. Morris[1] has recently reported another case.

That the "vestiges coccygiennes" of Tourneux and Herrmann and Mallory are the remains of the medullary canal there does not seem to be any doubt, and no attempt is made to deny the fact. It is even quite certain that in many cases they form a part of and give rise to the more complex congenital malformations, the tumors of monogerminal origin of this region. But it would certainly seem that here their significance ceases; they are to be regarded merely as a part of the original way of communication between the medullary canal and the skin surface, and are destined for obliteration. Nor have, in all probability, the connective-tissue strands of such importance to Stolper any causative relation with our dimples, sinuses, and cysts. If we are to regard the sinuses and cysts as more advanced forms of the dimple, as is undoubtedly the case, it is difficult to see how the dilated ends of these sinuses, and more particularly the sacculated pouches and the more extensive cyst formations, may be produced by traction of a few strands in this manner. And, too, would these binding strands tend to loosen as age advances, and when we know connective tissue thickens and becomes less mobile? We find, it will be remembered, our largest number of dimples in infancy and childhood.

It would seem that if the medullary remains were earlier or primary forms of our coccygeal defects, we should sometimes find in the latter evidences of the fact in the form of anomalies of the lining skin; the presence of cuboidal, columnar, or intermediate forms of cells approaching the squamous variety of the skin and transitions, one to the other. Likewise, in our medullary vestiges we might expect to find at times irregularities; portions of stratified squamous epithelium or transitional forms with or without persistence of the structure as a whole. Furthermore, as has been mentioned, nerve tissue is notably absent in the walls or in the tissue surrounding our sinuses and dermoid cysts. Histological examination shows us that in our epidermoid structures nothing but true skin has ever been found, except in those instances where it has not been possible to identify any lining tissue on account of inflammatory changes. As regards "the cells lining the central cavities of these structures (medullary vestiges), some become flat, resembling squamous epithelium (as that of the superficial skin), which might indicate a return to the former ectodermal type. Others become elongated and resemble cylindrical cells of the ependyma. The cells forming the mass of the medullary remnant retain their original spherical or polyhedral shape. Ciliated epithelium has not been found." Mallory mentions columnar and cuboidal epithelium. Tourneux and Herrmann found stratified epithelium of the prismatic type and pavement type. They approach either the cells of the medullary tube or those of the epidermis in their evolution. That the elements of the medullary vestiges, instead of atrophying, may take on a lawless growth, just as occurs in tissue with tumor formation elsewhere in the body, we have reason to believe, as has been said, because tumors of this region have been described containing cysts accompanied by or embedded in nervous or neuroglia tissue. Such are the cases of Gutzeit,[1] Weigert,[2] Wasle,[3] Perman, Schonborn, etc. The cysts in many cases were lined with cuboidal, cylindrical, and stratified squamous epithelium, often one form merging with the other. Such a case was Perman's. "The cysts lined with pavement epithelium not infrequently show the general character of a dermoid cyst; there may be a cornification of the epithelium (Hamel[4]); a development of papillæ (Nasse[5]); hair (Hamel, Lutkemuller,[6] Hildebrand[7]); and of sebaceous and sweat glands (Lutkemuller, Nasse, Hildebrand)."

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[1] Annals of Surgery, 1900.

It is not unusual to find fistulæ and dermoid cysts occurring in conjunction with these complex tumors of this region. Nor is it to be regarded as evidence against their formation from the ectoderm of the surface. Agglutinations and involutions in these cases occurring in similar manner as in uncomplicated cases, while other displacements of ectoderm, or part only of the same, together with medullary remnants and other fœtal structures by proliferation, form the bulk of the tumor mass. (See Schmidt's[8] case.) He found in a three-and-a-half-year-old girl a number of fistulæ in the sacrococcygeal region which communicated with a dermoid containing hair. This was connected to a complex tumor which had a pedicle connection with the spinal column.

Furthermore, we have to consider such cases occurring elsewhere; in the lumbar region or higher (cases of Muscatello,[9] Kaufmann,[10] Wette, etc.), where there are no medullary vestiges. We find, therefore, except in the large and progressively growing tumor formations on the one hand, medullary remnants, and upon the other, formations of true skin. There are no gradations in these smaller defects. So that it would seem that the epidermal lining of our dimples, sinuses, and cysts was derived from a tissue or cellular structure already differentiated and fixed and subject to no possibility of any other outcome. If this is not so, we will have to believe that the ectoderm of the distal portion of the tube becomes fully developed epidermis, or nothing at all, and disappears. If it is true that we have these dermoid structures, because the surface type of epithelium is developed, then we have a right at times to expect an irregularity in development or an irregular disposition of this epithelium. So far in many cases we have yet to see squamous epithelium of the surface in conjunction with cuboid, spherical, or polyhedral shaped and disposed cells. In the utter lawlessness prevailing in tumor formations in this region, however, this is to be expected, and is found.

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[1] Dissertation, Königsberg, 1896.

[2] Virchow's Archiv, 1875.

[3] Dissertation, Wierzburg, 1896.

[4] Dissertation, München, 1895.

[5] Langenbeck's Archiv, 1893-1895.

[6] Oesterr. med. Jahrb., 1875.

[7] Arch. f. klin. Chir., 1895.

[8] Dissertation, Greifswald, 1889.

[9] Archiv f. klin. Chir., 1894.

[10] Dissertation, Berlin, 1883-1893.

Again, we have to consider the cases where two, three, or even four congenital fistulous tracts are seen. It is difficult to see how the ascending end of the vestigial loop can account for these.

The fact that we can demonstrate dimples, sinuses, and cysts along the spinal column, in the neck, face, back of head, and median line, in mouth, etc., where there are no spinal cord remains, but where embryonal parts do meet and fuse, and where involutions or abnormal curvings and agglutinations may occur, seems certainly to establish an analogy in formation with those of the median line in the sacrococcygeal region. Cases of White,[1] Abraham,[2] Sutton, Wagstaffe,[3] etc., are referred to.

Further study of anatomical material upon the embryology of this region will form the basis of a subsequent paper.

Of the dermoid cysts, we speak of the true congenital dermoid lined with squamous epithelium and found usually upon the dorsal surface of the sacrum and coccyx, and which never reaches a size larger than that of the "fist," and "usually does not assume such size until later in life," and even then they are soft and can be "moulded" on pressure (Streit). Sometimes there is a fistulous communication with the skin. Their etiology is that of the depressions and sinuses met with in this region. Stolper considers them together with the dimples and sinuses, which he regards as the simplest forms of abnormality occurring as the result of "disturbance in the approximation of the embryonal component parts at the lower end of the axis skeleton." On the ventral side of the coccyx and sacrum, sometimes above, but oftener below, the levator ani diaphragm, we also find cystic tumors, "which must be classed as true dermoids." These are usually found of larger size than those of the dorsum. In Deahna's[1] case, pressure ulceration of the vagina and bladder actually happened, the cyst filling the pelvis. In Biernacki's,[2] a similar one prevented childbirth. They seldom, however, reach the size of the teratoid mixed tumors. Sanger[3] believes that this latter class are not truly congenital, but are present "simply as a germinal layer at birth," and must be considered a separate species of tumor. That dermoid cysts of this region are rare is shown by his collection of only eleven cases (three between rectum and coccyx, one between rectum and sacrum, three subperitoneal, in cavity of pelvis under levator ani, three behind rectum in cavity of pelvis, right and left side, one behind and below Douglas' cul-de-sac, but above recto-vaginal septum).

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[1] Transactions of the Pathological Society, London.

[2] Journal of Anatomy and Physiology, London, vol. xv.

[3] Transactions of the Pathological Society, London, 1878.

They are not visible as tumors, and are generally met "together with anomalies in the structure and differentiation of the uro-genital apparatus and the anus." They sometimes simulate ovarian or intraligamentary cysts or spina bifida with meningocele. (Death has resulted in two recorded instances from tapping the meningocele cysts mistaken for vaginal cysts and teratoid tumors.) They may rupture after suppuration into the bowel, and their contents—débris, pus, and hair—be passed by rectum. They may be mistaken for periproctitic abscesses. (Collected cases of Sanger, Meyer,[4] von Bergmann, Nasse, etc.)

These dermoid cysts are not confined to the regions immediately in front of and behind the sacrum and coccyx, but are found also in other regions of the pelvic connective tissue. They are noted for their simple construction, Sanger says, and show "no relation to the ovaries, the external skin, and spinal column." He distinguishes those situated high up and those that are low down. The former are more complex structures, generally containing mesodermal and ectodermal tissue, and, therefore, hardly belong to the class of simple, true dermoid cysts, but rather to the teratoid class.

To return to the simple cystic structures of true dermoid character. "The simplest forms of these disturbances, which at the same time tend to throw light on the origin of the larger real dermoid cysts, occur in the form of flat depressions in the skin in the region of the coccyx, or as fistulæ in the same region." Frequently at the end of these fistulæ coccygeæ, lined with epidermis and often containing hair and sweat glands, we find dilatations or cavities, or "a series of epidermoidal sacs (Schmidt[5]) containing atheromatous material, and which may without

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[1] Archiv f. Gynäkol., 1876, vol. vii.

[2] Dissertation, Berlin, 1887.

[3] Archiv f. Gynäkol., 1890.

[4] Dissertation, Greifswald, 1890.

[5] Virchow's Archiv, 1888; Arb. aus der Chir. Poliklin. zu Leipzig.

doubt be considered as dermoid cysts formed from the segmented or strangulated ends of such fistulæ;" also these epidermoid fistulæ themselves may, owing to closure of their orifices, dilate and then resemble, and, in fact, constitute, as has been said, dermoid cysts. These cysts may or may not present a dimple or sinus superficially. This character of dermoid is noted for its very slow growth. In this connection the case of Kutz[1] is of interest. He mentions a certain tumor, the size of two fists, situated upon the posterior surface of the sacrum, in a woman, aged fifty-three years, and which was noticeable only as a small nodule at birth. (Cases, also, of Lannelongue, etc.) The simple dermoids of this nature situated upon the ventral surface possess the same character, and may or may not have a dimple or fistulous opening at this site. Usually there is nothing to suggest their presence externally. They are, as a rule, situated between the anterior surfaces of the sacrum and coccyx and the rectum in the loose connective tissue. Birkett's[2] case was probably one of this kind; Sanger[3] classifies it as such, although histological data are scanty. The origin of these cysts is explained upon somewhat different grounds than those given for the structures on the dorsum and probably formed by agglutination of the ectoderm with the skeleton. They stand in very intimate relation "to the cloaca formations and the secondary processes resulting therefrom." Born believes that during and succeeding the cloaca stage, with the processes of invagination and fusions that happen, epithelial germinal cells can become separated and subsequently develop cysts. Aschoff thinks it not astonishing that we find here cysts lined with epidermis or cylindrical epithelium, as the cells of the ectodermal plate are capable of producing epidermis as well as mucous membrane. He instances in this connection the "manifold character" of the urethral mucous membrane which is derived from the urogenital plate. Also, the ectodermal structures concerned in the formation of the cloaca may be used in explanation of the cysts lined with mucous membrane found here. There is, as has been demonstrated (Spee[4]), a "direct transition of entodermal structures into offshoots of the ectoderm" through the normally early obliterated canalis neurentericus, connecting the hind end of the medullary canal and the post-anal intestine. Borst speaks of cysts lined partly with squamous epithelium in this region, which he considers formed from this remnant.

The complex tumors of this ventral region also present cysts lined with stratified, squamous epithelium and mucous membrane and transitional varieties. True dermoid cysts are found forming a part of these complex masses or next to tumors of complex construction. Borst, also, while thoroughly doubting Ritschl's view of the derivation from the coccygeal vestiges, says that inasmuch as we have sometimes such great disturbances in this region, possibly in stunted conditions of the lower end of the axis skeleton, " portions of the vestiges coccygiennes may be transposed to the ventral side and give rise to tumors."

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[1] Dissertation, München, 1895.

[2] Guy's Hospital Reports, 1859.

[3] Berl. klin. Woch., 1884.

[4] Archiv f. Anat. und Phys., 1889.

We find transitions from the simple congenital dermoid cyst to the larger and more complex dermoids, and from these to the simpler mixed or teratoid tumors monogerminal in origin, and from these again to the elaborately complex masses due to a secondary germinal implantation or true fœtal inclusion.

We may regard the foveæ coccygeæ as the simplest of the abnormal disturbances and the mixed teratoid tumors the most complex that occur in this region. All the transition forms are observed. Both Hamel and Masse saw stratified pavement epithelium merge into the ciliated form. Borst and Hildebrand saw cysts that were lined with epithelium resembling that of the intestine, with a basilar border and mucous glands, in which there was a transition to stratified pavement epithelium.

The tumors situated upon the dorsum of the sacrum and coccyx show more or less firm connection with the bone beneath, while those upon the ventral surface have but slight connection, if at all, occasionally being attached to the periosteum. This is what one would expect to find, considering our ideas of origin. That inflammatory changes are prone to occur in the deeper of these structures, the fistulæ, in the pouch-like dilatations often found at their terminations and in the cysts, we have seen. The sebaceous accumulations, proliferated epithelium, and especially hair, products of the lining epidermis, having no outlet, or, at best, a greatly narrowed one, are apt to be retained and lead to irritation and suppurative inflammation. If there be a pre-existing outlet, the pressure of the retained material may re-open it and the result be a persistent chronic or intermittent discharge of pus and débris. If none existed, or the pressure of the accumulation be insufficient to open it, having become sealed through maceration of the skin at the outlet, or if the opening show great disproportion in size, retention and abscess formation result, with the usual consequences: tumor formation with subsequent burrowing of pus and the establishment of secondary fistulous tracts. These fistulous tracts are always to one side of the median line, never in it.

Traumatism of one nature or another is very often the exciting cause for inflammatory change. A blow or nothing more severe than a long period of rest in bed may be sufficient. Wette found seven cases in 15,000 sick persons. He also mentions a case occurring subsequent to the formation of a bed-sore in a typhoid patient. Stolper's first case followed confinement to bed (typhoid). Direct traumatism is, however, more often the exciting cause, as in the case of Routier and

Dolens,[1] Goodsall,[2] Anderson,[3] J. M. Warren,[4] etc. Abscess formation followed in periods of time varying from a few weeks to several months or years. The *secondary* sinuses were situated at points close to the median line, well out on the buttock, and from near the sacro-iliac synchondrosis to the upper posterior and inner surface of the thigh on either side. (Goodsall, in the analysis of his cases, thinks the first rupture occurs, usually from one-quarter of an inch to one-half of an inch on the left side of the median line.) Furthermore, these discharging sinuses have frequently been considered to have their origin in a focus of *tubercular disease* occurring in the sacrum or coccyx, with the result that one or the other of these bones has been curetted nearly away. The cases of Beall,[5] Goodsall, Madelung,[6] Gussenbauer[7] and others contain such examples.

The treatment that these cases have usually received has been incision of the abscess formation and curetting of the discharging sinus, followed by the use of carbolic acid or silver nitrate as caustics to destroy the lining membrane or to facilitate healing. The only plan, however, promising complete success is the radical, complete extirpation of the whole tract down to the periosteum of the bones as soon as practicable.

A CASE OF TRANSITORY CYSTINURIA ASSOCIATED WITH DIAMINURIA.

By Milton W. Lewis, M.D.,
AND
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OF BALTIMORE.

In a previous paper on cystinuria one of us collected all the cases reported up to midsummer, 1899. Including Simon's[8] own case, the number was then 107. Since that time three additional cases have been reported by Warburg,[9] Cammidge and Garrod,[10] and Reid.[11] We

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[1] Bull. et Mém. Soc. de Chir., 1888.

[2] St. Bartholomew's Hospital Reports, 1858.

[3] Boston Medical and Surgical Journal, 1847.

[4] The American Journal of the Medical Sciences, 1854; Philadelphia Medical Times, 1873; Boston Medical and Surgical Journal, 1877.

[5] Atlanta Medical and Surgical Journal, 1889.

[6] Centralbl. f. Chir., 1885.

[7] Prag. med. Woch., 1893.

[8] Cystinuria and its Relation to Diaminuria. The American Journal of the Medical Sciences, 1900, vol. cxix.

[9] Ueber Cystinuria. Deut. med. Zeit., 1898; cit. in Centralbl. f. d. med. Wiss., 1899, No. 13, p. 222.

[10] On the Excretion of Diamins in Cystinuria. Journal of Pathology and Bacteriology, 1900, p. 327.

[11] A Case of Cystinuria Ending in Recovery. New York Medical Journal, 1901, vol. lxxiii, No. 16.